

EVALUATION OF THE IMPACT OF A WEB-BASED EDUCATIONAL TOOL ON AWARENESS OF NEWBORN SCREENING AND CARRIER TESTING

by

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ABSTRACT

Introduction: There is considerable lack of awareness of newborn screening (NBS) among patients in the prenatal setting. Only about 20 states have designed specific educational materials on NBS that are distributed during pregnancy. Also, previous studies have shown that African American women receiving prenatal care believe that screening for sickle cell disease is beneficial, but they do not personally find themselves at an increased risk to have a child with sickle cell disease. To increase awareness of newborn screening and carrier screening for sickle cell disease, cystic fibrosis, and the thalassemias, we developed a website called My Baby's Health. This website provides education on NBS and carrier screening that is tailored to the patient's ethnicity. The goal of this study is to evaluate this method of educating pregnant women on newborn screening and carrier testing.

Methods: Women in their 1st or 2nd trimester of pregnancy were approached to access the My Baby's Health website on a computer kiosk at the clinic. They were encouraged to take brief surveys before and after reading the information on the site. The pre-website survey asked questions on the patient's previous knowledge of sickle cell, cystic fibrosis, and the thalassemias, carrier testing, how these conditions are inherited, and how newborn screening is performed. The follow-up survey asked the same knowledge-based questions on the genetic conditions and newborn screening, as well as questions on the participant's opinion of the site.

Results: Twenty-five participants completed both pre-and post-website surveys. Knowledge of NBS and carrier testing did improve on the post-test, and all individuals found the website at least somewhat helpful.

Conclusion: The website is helpful in increasing knowledge of sickle cell disease, and all participants found it at least somewhat useful. However, one of the main challenges is implementing this website into the workflow of a clinic so that it has maximum benefit. Using educational tools like this website may have a public health benefit by decreasing disparities in NBS services across the United States, since lack of awareness can lead to anxiety and failure to comply with recommendations for follow-up.

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1.0 INTRODUCTION

There is a significant lack of awareness of newborn screening in the United States [1]. To remedy this knowledge gap, the Genetic Alliance launched a website in September 2011 called “Baby’s First Test” (www.babysfirsttest.org), which gives patient-friendly information on the genetic conditions included on the newborn screening panel for each state. This website also describes what to expect during the newborn screening process and provides resources in the event a baby tests positive for a condition. With the support of Genetic Alliance, the website “My Baby’s Health” was developed to serve as a patient resource during the prenatal period. The goal of this project was to incorporate this website into the flow of a prenatal clinic and assess its efficacy in educating the prenatal population on newborn screening and carrier testing.

1.1 MY BABY’ S HEALTH

The My Baby’s Health website (www.mybabyshealth.org) is an educational tool that gives basic information on genetics, newborn screening and carrier testing for sickle cell disease, cystic fibrosis, and the thalassemias. The first pages of the website describe the function of genes and how genetic testing is performed. The participant is then able to select as many ethnic backgrounds as she identifies with (African American, Asian, Caucasian, Hispanic or Southeast Asian), and the website will provide information on screening for the genetic conditions most

commonly associated with that population. Those of Caucasian ancestry receive information on cystic fibrosis, and all other ethnicities are presented information on sickle cell disease, alpha thalassemia, beta thalassemia, and cystic fibrosis. Clinical features of the genetic conditions, what it means to be a carrier and autosomal recessive inheritance are all described. At the conclusion of the website, a basic overview of newborn screening is reviewed, describing how the test is performed and how the results are handled. There is also a link to “Baby’s First Test”, so that viewers can look for more information specific to their state, including the specific genetic conditions included on that state’s panel.

1.2 SPECIFIC AIM 1

The first aim of this project was to integrate the “My Baby’s Health” website into the workflow of the prenatal clinic.

1.3 SPECIFIC AIM 2

The second aim of this project was to evaluate the impact of the “My Baby’s Health” website in educating pregnant women on the basics of newborn screening and carrier testing.

2.0 BACKGROUND AND SIGNIFICANCE

2.1 NEWBORN SCREENING

2.1.1 Overview

Newborn screening is a mandatory public health program that began in the early 1960's to identify genetic conditions that pose a significant health risk if left untreated and for which there is a treatment option available [2]. Phenylketonuria (PKU) was the first genetic condition to be screened for after Robert Guthrie developed the bacterial inhibition assay to measure blood phenylalanine levels as well as the filter paper for the blood spot test [3]. Over the years, newborn screening has expanded to include a wider range of genetic conditions. This testing is ideally conducted between 48-72 hours after birth by obtaining a blood sample through a heel stick. A hearing test is also part of the newborn screening process.

All states in the United States perform newborn screening, but the specific regulations and the conditions included on the panel vary by state. In 2006, the American College of Medical Genetics (ACMG) issued a statement recommending 29 genetic conditions that should be included on every state's newborn screening panel [4]. This Recommended Universal Screening Program (RUSP) is composed of conditions in the following categories: hemoglobinopathies, inborn errors of organic acid metabolism, fatty acid oxidation disorders, amino acid disorders,

and other miscellaneous diseases including congenital hypothyroidism and cystic fibrosis [2]. States are not required to abide by these recommendations but can use them to inform their own NBS practices. Since that time, the RUSP has expanded and includes 31 core conditions and 26 secondary conditions as of December of 2012. In order for a disorder to qualify for inclusion as a core condition on the RUSP, testing should be feasible in 24-48 hours after birth, have a treatment, and have a known natural history. It has become a key component of preventive pediatric medicine [5].

2.1.2 Educational gap

Since newborn screening is mandatory, there is a concern for a lack of parental education. When consent is required for a medical test, providers are required to, at the very least, inform parents that the test is being done and to obtain their permission. The fact that NBS is mandatory does not mean the same level of parental education is not necessary, however data have shown that NBS is often not discussed with parents [3].

It is well understood that parental education of newborn screening is essential, and most states have some type of educational measures in place [6]. Brochures outlining basic information are a common method of education. Newborn screening is a complex system, and parents should be made aware of the basic procedures, significance of testing, possible outcomes, and the need for follow-up with a positive result [7].

Davis et al discussed the findings of a focus group of parents and providers, which indicated that parents had very little familiarity with newborn screening [8]. Almost no parents had heard the term “newborn screening”, though some recalled a “heel stick test.” Some parents had heard of PKU, but were unaware that newborn screening tested for other genetic conditions

as well. Most parents did not recall being given educational materials on newborn screening during the prenatal period. Many said that they were given brochures after delivery, but few actually read them. [8]

Bridging this educational gap is essential, as it has been suggested that if parents are aware of the purpose and process of screening, they may act more promptly if their child tests positive [9]. Additionally, if parents have been informed that a positive test result is not diagnostic and requires confirmatory testing, it may help lessen the stress of a false-positive result. Tluczek et al found that parents whose newborn had an abnormal NBS result for cystic fibrosis had higher anxiety if they had less knowledge of newborn screening [10]. Finally, open communication on the process of newborn screening is important to promote confidence in this program as a public health initiative, particularly because there has been recent debate over the use of remaining blood spot samples for research purposes [9].

2.1.3 Integrating education into prenatal period

Approximately 20 states require the distribution of newborn screening educational materials during the prenatal period [6]. However, in many cases, materials are distributed at inopportune times, such as after delivery [8]. The time period after delivery, when parents are exhausted and focused on the immediate needs of the newborn, is not optimal for a discussion on newborn screening. There has also been evidence for a disparity in education depending on socioeconomic status. Tluczek et al found that mothers with a lower income were more likely to receive newborn screening information after delivery than those with a higher income [11].

Many studies have suggested that newborn screening is best discussed prenatally, but it is frequently not explained during that time period [1, 7, 8, 12]. Faulkner et al. found that only

33% of prenatal care providers discussed newborn screening with their patients [1]. Common factors limiting this discussion were that prenatal providers believed that pediatricians and other hospital staff would be the ones to explain newborn screening or that patients never inquired about it. Hayeems et al. found that providers who felt a responsibility to discuss NBS with patients were three times more likely to do so, and those who lacked the confidence to counsel on NBS were 70% less likely to do so [12]. This data suggests that an educational tool, such as the My Baby's Health website, containing all the pertinent information on NBS could help remove some barriers to patient education and help providers feel more equipped to discuss screening with their patients. Having patients view the material during their clinic visit could take some of the responsibility off of the prenatal providers when they meet with patients, especially those who do not feel confident enough to explain NBS.

2.2 CARRIER TESTING

2.2.1 Sickle cell disease

Sickle cell disease (SCD) is an inherited disorder characterized by the production of sickled hemoglobin. This condition is caused by bi-allelic beta-S mutations in the HBB gene, which codes for the production of beta-globin [13]. Those with a mutation on only one allele are considered to have sickle cell trait (SCT). Sickle cell disease can also occur if a beta-S mutation is inherited from one parent and a different beta-globin mutation is inherited from the other parent. The most common examples of this compound heterozygosity are sickle cell-hemoglobin C disease and sickle beta-thalassemia. Common features in affected individuals

include severe hemolytic anemia, pain crises, suppressed immune system, stroke, and organ/tissue damage, especially of the lungs, bones and kidneys [13]. This condition is particularly prevalent in those of African, South American, Central American, Saudi Arabian, Indian, and Mediterranean descent [14]. The Center for Disease Control (CDC) estimates that between 90,000 and 100,000 Americans are affected by SCD, and it occurs in 1 out of every 500 Black or African-American births [14]. Approximately 1 in 12 African Americans has sickle cell trait [14]. Testing to determine if someone has SCT is performed using hemoglobin electrophoresis, which detects variations in types of hemoglobin in the bloodstream.

2.2.2 Cystic Fibrosis

Cystic Fibrosis (CF) is a multi-system disease that primarily affects the epithelial cells of the respiratory tract, hepatobiliary system, pancreas, intestine, and male genital tract [15]. It is an autosomal recessive condition caused by mutations in the *CFTR* gene, which controls the chloride channels of a cell. Improvements in treatment have increased the life expectancy of someone with cystic fibrosis to be about 37 years [15]. Cystic fibrosis is among the first genetic conditions to have a screening test for carrier status in the general population. It is most common in those of Caucasian or Ashkenazi Jewish descent, with carrier frequencies of 1:28 and 1:29 respectively [15]. The ACMG has published screening guidelines that recommend screening the general population with a panel of 23 *CFTR* mutations [16].

2.2.3 Alpha Thalassemia

Alpha thalassemia is an autosomal recessive hemoglobinopathy that causes microcytic hypochromic anemia [17]. It is most prevalent in those of Mediterranean, South-East Asian, African, Middle Eastern, and Indian ancestry [18]. There are two clinically significant presentations: hemoglobin Bart hydrops fetalis (Hb Bart syndrome) and hemoglobin H disease. Hb Bart syndrome is the most severe form of alpha thalassemia and typically is fatal in the neonatal period. Hemoglobin H disease is typically associated with anemia, mild jaundice, hepatosplenomegaly, and some bone abnormalities [17].

2.2.4 Beta Thalassemia

Beta thalassemia is a blood condition characterized by reduced production of hemoglobin, causing microcytic hypochromic anemia [19]. There are two main classifications: major and minor thalassemia. Thalassemia major usually presents within the first 2 years of life, with symptoms including failure to thrive, jaundice, and enlarged spleen and liver. This condition can also cause bone deformity and delayed puberty. Thalassemia intermedia is less severe and typically manifests later in life. Symptoms can include anemia, bone changes, and hepatosplenomegaly. Beta thalassemia occurs most frequently in those from Mediterranean countries, North Africa, the Middle East, Indian, Central Asia, and Southeast Asia.

2.2.5 Perceptions of sickle cell disease and carrier testing

Previous studies have suggested that the African American community perceives sickle cell as a serious disease, but individuals generally do not believe they have a significant risk to have a child with that condition. Further, there is relatively low uptake of education of sickle cell disease. Some barriers to this education include a desire for avoidance, since some believe that not thinking about genetic conditions makes it less likely for them to occur [20]. Long and colleagues conducted a focus group of African American individuals to elucidate their perception of SCD and SCT, as well as carrier testing and newborn screening [20]. That study found that there is a perceived benefit to carrier testing and newborn screening, because of the value in being aware of a child having a medical condition in advance and having the option to choose whether to continue a pregnancy.

In the African American community, it is common to rely on personal or secondhand experiences when understanding the genetics of sickle cell disease [20]. Using friends and family as the primary source of information increases the chance of being misinformed. Further, it has also been shown that African-American women were 50-70% less likely to use health information resources such as news media and computers [21]. Increasing the utilization of health resources, such as websites like My Baby's Health, can reduce the likelihood of individuals being misinformed about genetics and specific genetic conditions.

2.2.6 Benefits of prenatal education on carrier testing

As with newborn screening, education on carrier testing has been suggested to improve follow-up if a carrier test comes back positive. Generally, the follow-up rate for those with sickle cell disease, sickle cell trait, and other hemoglobinopathies ranges between 35 to 60 percent in the United States [22]. Potential factors that hinder follow-up include anticipatory anxiety, guilt, and denial of having a child with a health problem. It has been shown that education during the prenatal period improves follow-up for those with sickle cell trait [22].

2.3 PATIENT EDUCATION

2.3.1 Computer Education

Computer education has emerged as a way to make medical information more accessible to patients in a more cost-effective way. These computer programs can be more interactive than paper materials, allowing patients to have stronger improvement in knowledge and to have more involvement in medical decision-making [23]. Individuals have differing baseline levels of knowledge, so it can be challenging to develop a program that is appropriate for a wide range of people. Multiple studies have found that those from rural areas and a lower socioeconomic status tend to respond well to computer-based education, suggesting that the My Baby's Health website could be a useful tool to educate that population [23]. Learning from a website allows individuals to read through the information at their own pace, which can be beneficial for those with lower literacy skills. Keulers et al found that retained knowledge may be even higher for

those who were educated through a computer rather than by a provider, and that patient satisfaction was equal for both methods [24].

3.0 METHODS AND PROCEDURES

3.1 PARTICIPANT RECRUITMENT

Participants were recruited from the Outpatient Clinic at Magee-Women's Hospital of University of Pittsburgh Medical Center from August 2012 to February 2013. Women who were in their first or second trimester of pregnancy at their first obstetrical visit were eligible. They were approached after having their blood work drawn at the end of their clinic visit. They were taken to a computer kiosk in the clinic with a link to the My Baby's Health website. Exclusion criteria included women who were under the age of 18, did not speak or read English, or were incarcerated.

3.2 CONSENT

Before taking the surveys, participants can read through a paragraph describing the goal of the study. The paragraph states that their participation is voluntary and that they may withdraw at any time. There were no foreseeable risks to completing the surveys. Contact information for study personnel was provided. See Appendix A for the consent paragraph.

3.3 COMPENSATION

Participants were offered a free water bottle after viewing the website if they completed both surveys.

3.4 PRE-AND POST-WEBSITE SURVEYS

The pre-website survey was composed of one question asking which genetic conditions the participant had heard of, six knowledge-based questions and six questions on demographic information. The post-website survey had the same six knowledge-based questions, as well as five opinion-based questions on the helpfulness of the website. Surveys were taken anonymously. See Appendix A for the survey questions.

3.5 STATISTICAL ANALYSIS

The knowledge-based portion of the survey results was analyzed using non-parametric tests to determine if participants had improved performance on the post-website test. Non-parametric tests were used under the assumption that the data would not follow a normal distribution. Test scores were also compared with age and number of other children to evaluate the effect of those factors. Finally, the data were stratified by race to determine if there is any evidence suggestive of culture bias in the website. Other demographic data including education level and relationship status were also evaluated in their relationship with test scores.

4.0 RESULTS

A total of 34 women took the pre-website survey, and 31 took the post-website survey. Twenty-five took both surveys. Statistical analyses were conducted only on those who completed both surveys.

4.1 PATIENT POPULATION DEMOGRAPHICS

Out of the 25 participants who took both surveys, the age range was from 18-36, with a mean age of 22.8 years. Figures 1-4 below describes the educational background, relationship status, number of previous children, and ethnic background of those who completed the surveys.

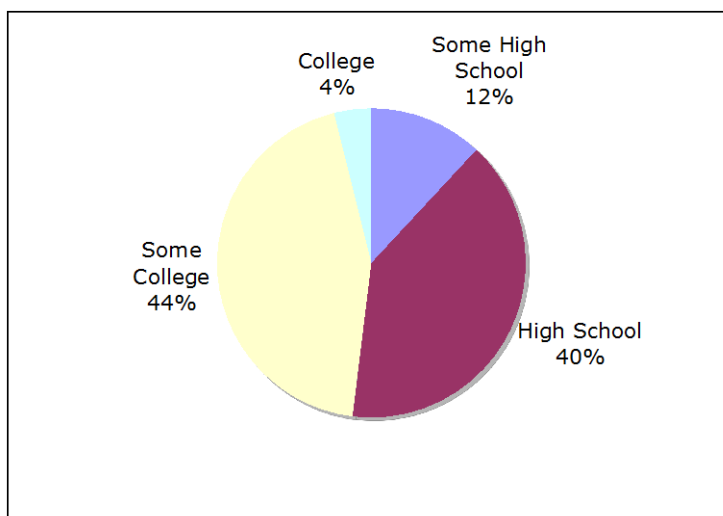


Figure 1. Participants' Educational Background

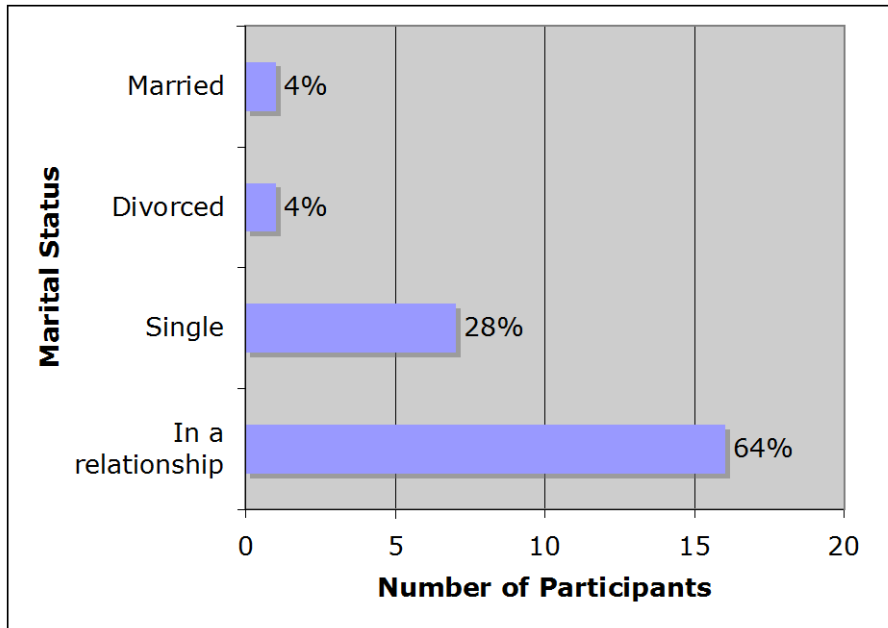


Figure 2. Study Population Relationship Status

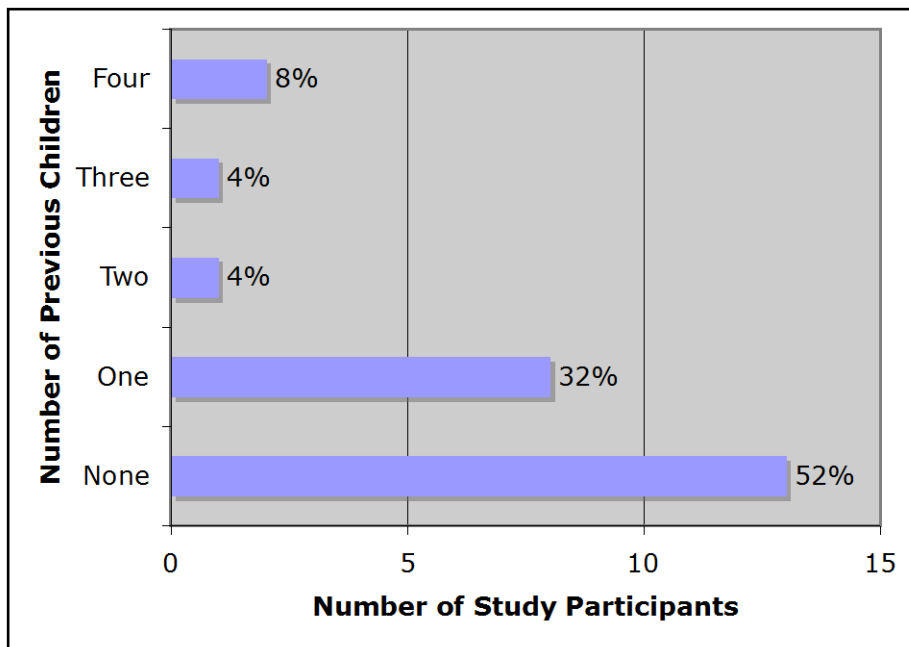


Figure 3. Study Participants' Number of Prior Children

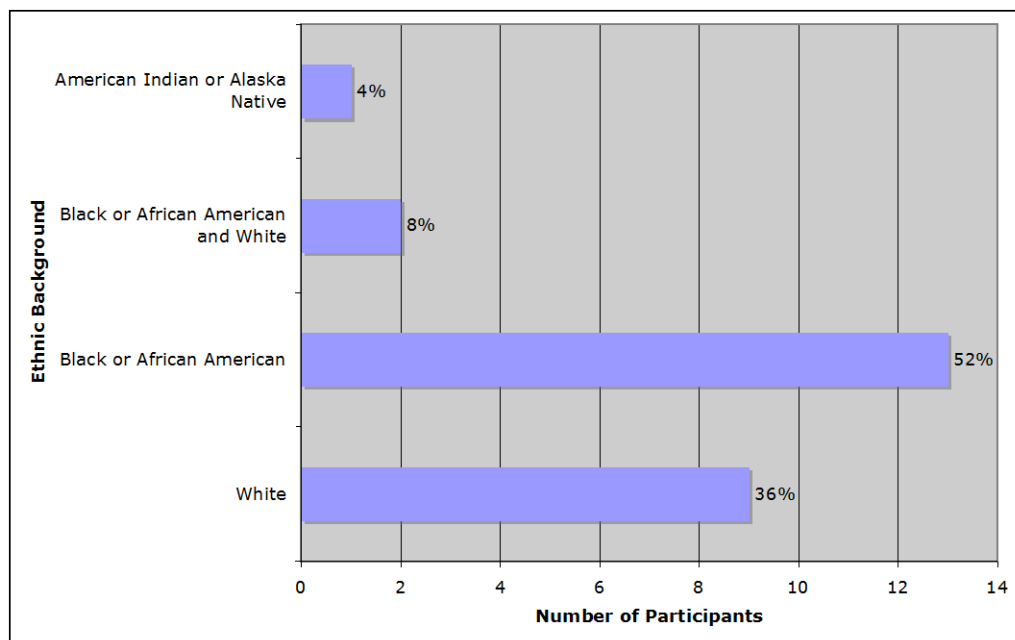


Figure 4. Participants' Ethnic Background

4.2 PRE- AND POST-WEBSITE SURVEYS

4.2.1 Prior Familiarity with Genetic Conditions and NBS

Figure 5 below depicts the participants' familiarity with sickle cell trait and disease, the thalassemias, cystic fibrosis, and newborn screening prior to viewing the website. Thalassemia was the condition with which participants were least familiar (36%), and there was the most prior familiarity with sickle cell disease (68%). Over half (60%) of participants had heard of newborn screening.

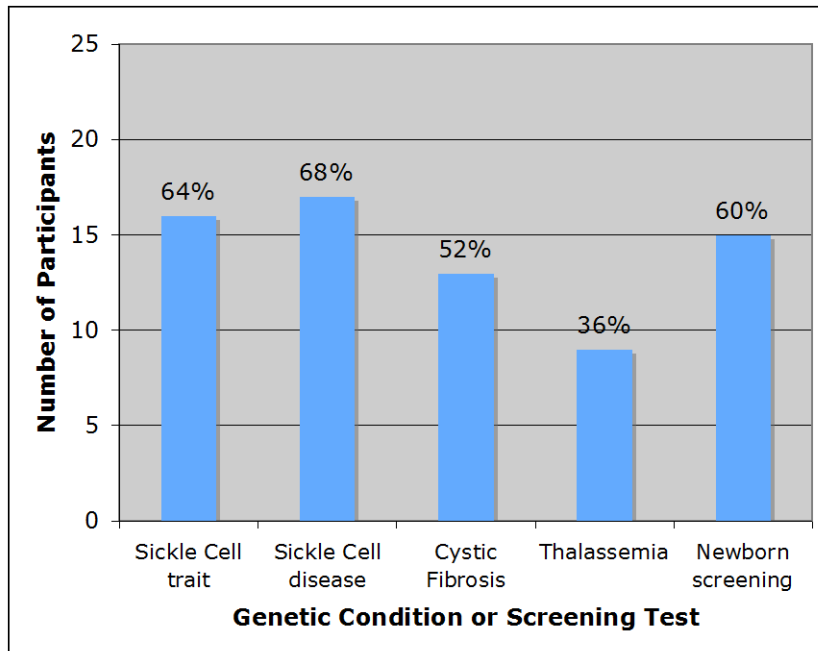


Figure 5. Participants Prior Familiarity with Genetic Conditions and Newborn Screening

4.2.2 Post-test vs. Pre-test Performance

To compare scores on pre- and post-tests, the Wilcoxon test for paired data was performed with a one-sided p-value. The null hypothesis was that there would be no difference in performance between the two surveys, with the alternative hypothesis that scores on the post-test would be improved over the pre-test. Scores were shown to be higher on the post-test, with a total p-value of 0.000667. When divided into individual questions, there was statistically significant improvement on questions 1, 2, 4, and 5. The results of this analysis are summarized in Table 1 below.

Table 1. Pre-Test and Post-Test Performance (n=25)

Survey Question	Correct Responses on Pre-Website Survey	Correct Responses on Post-Website Survey	P-Value
What does a positive sickle cell carrier test mean?	8 (32%)	16 (64%)	0.01176
How can a child get SCD?	7 (28%)	18 (72%)	0.001301
What does a negative CF carrier test mean?	6 (24%)	10 (40%)	0.09083
T/F: Two parents who are CF carriers can have healthy children	9 (36%)	17 (68%)	0.006712
How is NBS performed?	10 (40%)	22 (88%)	0.000314
T/F: A healthy baby can receive an abnormal NBS result	10 (40%)	15 (60%)	0.07252
Average total score	2	4	0.0000677

McNemar's chi square test for paired data was conducted to evaluate each test question individually. The null hypothesis for this two-sided test was that there would be no difference between performances on each test, while the alternative was that there was a difference (not necessarily an improvement). This test also found a statistically significant difference in scores on questions 1, 2, 4, and 5. The p-values are listed in Table 2 below.

Table 2. Change from pre-test to post-test

Question Number	P-Value
1	0.04331
2	0.005546
3	0.2888
4	0.02686
5	0.001496
6	0.2278

Both the Wilcoxon and McNemar test were used because of the individual strengths and weaknesses of these tests. Wilcoxon allows a one-sided test, which provides higher statistical power for the question at hand since we are specifically looking for an improvement in the post-test over the pre-test score. However, with this test the symmetry assumption of the distribution may not necessarily be upheld. McNemar's test is ideal for testing independence of paired binary variables, but this can only be a two-sided test and would therefore have reduced statistical power.

4.2.3 Test Performance vs. Age

Using the Spearman correlation between pre- and post-test total score and age, it was found that age had no correlation with the performance on the pre-test. However, there was a statistically significant relationship between age and the score on the post-test. There was an inverse relationship, where the score on the post-test decreased as the participant's age increased. These findings are shown in Table 3 and Figures 6 and 7.

Table 3. Age vs. Test Performance

Test	Rho	P-Value
Pre-test total	-0.082956	0.6934
Post-test total	-0.525623	0.006967
Change (post-test total – pre-test total)	-0.42421	.03456

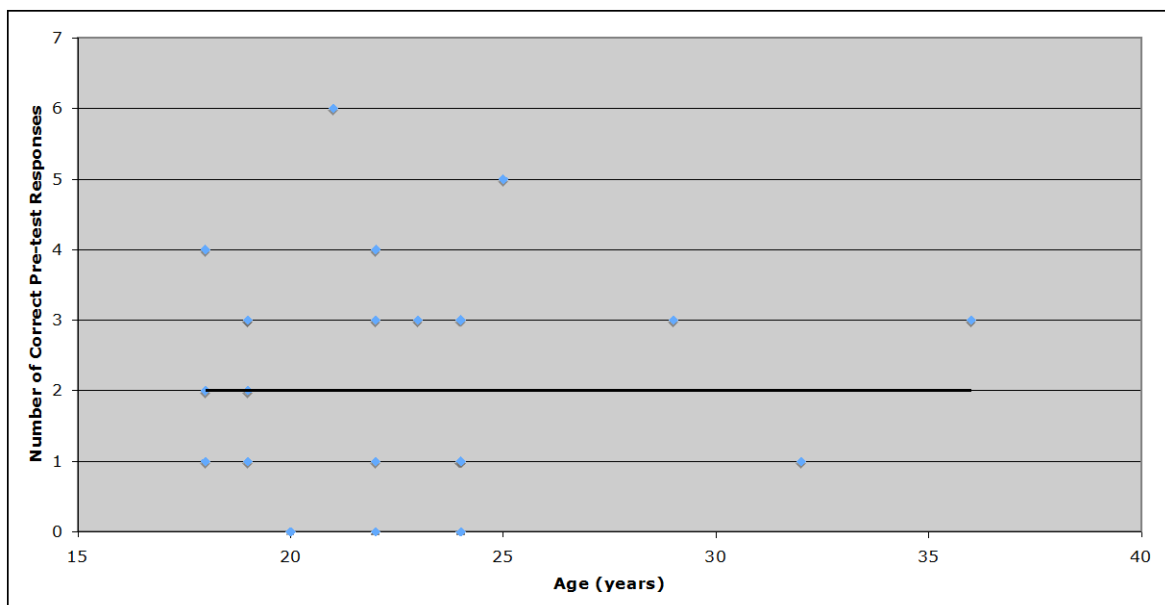


Figure 6. Pre-test vs. Age

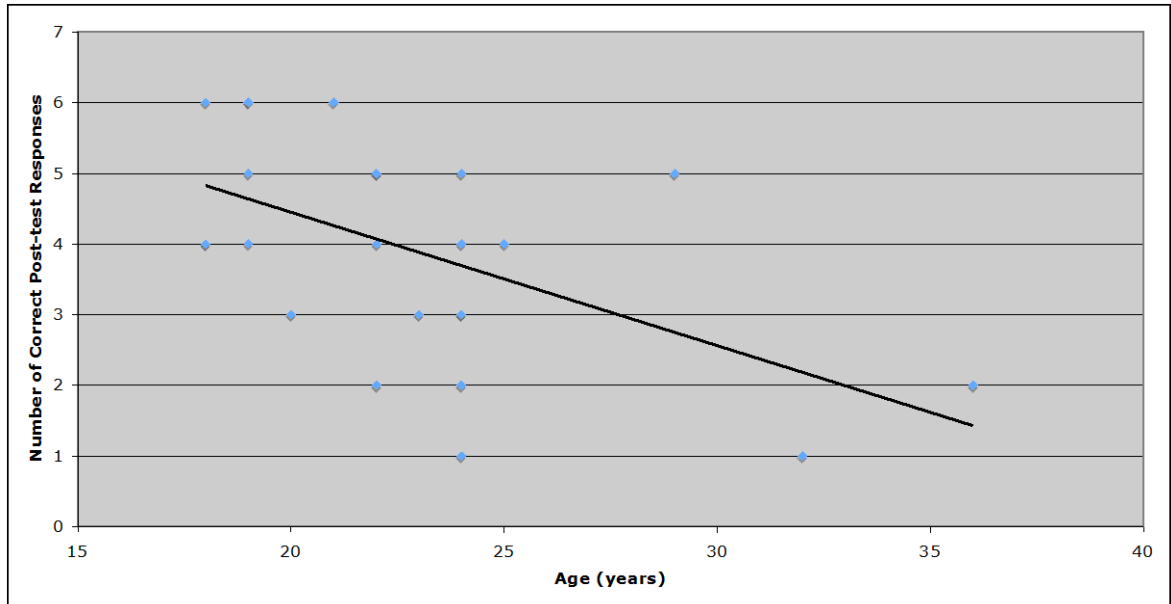


Figure 7. Post Test vs. Age

To evaluate if the number of other children influenced this age effect, the Spearman correlation was re-calculated adjusting for previous children. Table 4 contains the analysis while adjusting for either having or not having any previous children. Table 5 adjusts for the number (1-4) of previous children. The age effect was still present even when accounting for having other children.

Table 4. Age vs. Test Performance (adjusted for having had previous children)

Test	Rho	P-Value
Pre-test total	-0.29971	0.1455
Post-test total	-0.47762	0.01575
Change (post-test total – pre-test total)	-0.31627	0.1235

Table 5. Age vs. Test Performance (adjusted for the number of previous children)

Test	Rho	P-Value
Pre-test total	-0.18097	0.3867
Post-test total	-0.60918	0.001229
Change (post-test total – pre-test total)	-0.43435	0.03004

4.2.4 Test Performance vs. Having Previous Children

The effect of having had prior children was measured against pre- and post-test performance using the Mann-Whitney test. Those who claimed to have one or more previous children were compared with those who have never had a child. The analysis showed that those with at least one other child did better on the pre-test, but there was no difference in post-test performance. The results are listed in Table 6 below.

Table 6. Previous Children vs. Test Performance

Prior Children	Test	P-Value
Previous children vs. none	Pre-Test	0.04767
	Post-Test	0.9338
	Change	0.02677

4.2.5 Test Performance and Other Demographic Factors

Pre-and Post-test performances were compared with other demographics including race, educational background, and relationship status. The impact of ethnicity was analyzed using the Mann-Whitney test with a two-side p-value. For ethnicity, almost all individuals identified themselves as either African American or Caucasian. There were two individuals who called themselves both Caucasian and African American, as well as one person who was American Indian/Alaska Native. Race was analyzed in two ways, which differed in the way participants who were biracial or American Indian/Alaska Native were treated. In the first analysis, those who considered themselves only Caucasian were compared to all other ethnicities. Second, those who identified as African American, even if they also selected another ethnicity, were compared with those who did not say they were African American. There was no statistically significant difference in scores between ethnic groups. There was a nearly significant difference ($p=0.06984$) in the pre-test score between African Americans and non-African Americans, so it is possible that there is a race difference in knowledge before reading the website. There was no evidence for a difference in post-test performance. Therefore, if race is associated with differences in previous awareness of genetic conditions and newborn screening, the website may make up for any deficiency in knowledge. Table 7 below summarizes these findings.

Table 7. Race vs. Test Performance

Race	Test	P-Value
Caucasian vs. not Caucasian	Pre-Test	0.2456
	Post-Test	0.7952
	Change	0.1666
African American vs. not African American	Pre-Test	0.06984
	Post-Test	0.8246
	Change	0.1749

The scores of those in a relationship (stating they were either in a relationship or married) were compared to those who were not in a relationship (single or divorced). There was no difference in scores between the two groups, as shown in Table 8.

Table 8. Relationship Status vs. Test Performance

Relationship Status	Test	P-Value
In a relationship/married vs. single	Pre-Test	0.7652
	Post-Test	0.09656
	Change	0.4234

The role of a participant's educational background was compared to test performance to determine if those with a college background (either completing some college or having a college degree) scored differently than those who did not attend college at all (some high school or a high school degree). There was no statistically significant difference in test scores, as shown by Table 9.

Table 9. Educational Background vs. Test Performance

Educational Background	Test	P-Value
College vs. none	Pre-Test	0.2528
	Post-Test	0.618
	Change	0.3057

4.2.6 Opinion-based Questions

The following graphs (figures 8-11) describe the participants' feedback on the website, which was overwhelmingly positive. The vast majority of individuals (92%) said the website provided just the right amount of information. Additionally, 64% of participants found the information to be clearly presented, found it helpful in understanding genetic testing and NBS, and believed it better prepared them to discuss that testing with their prenatal care providers. The remainder of participants found the website to be somewhat clear and somewhat helpful in learning the information. Finally, 76% of participants would recommend the website to other women, and the remainder would consider recommending it.

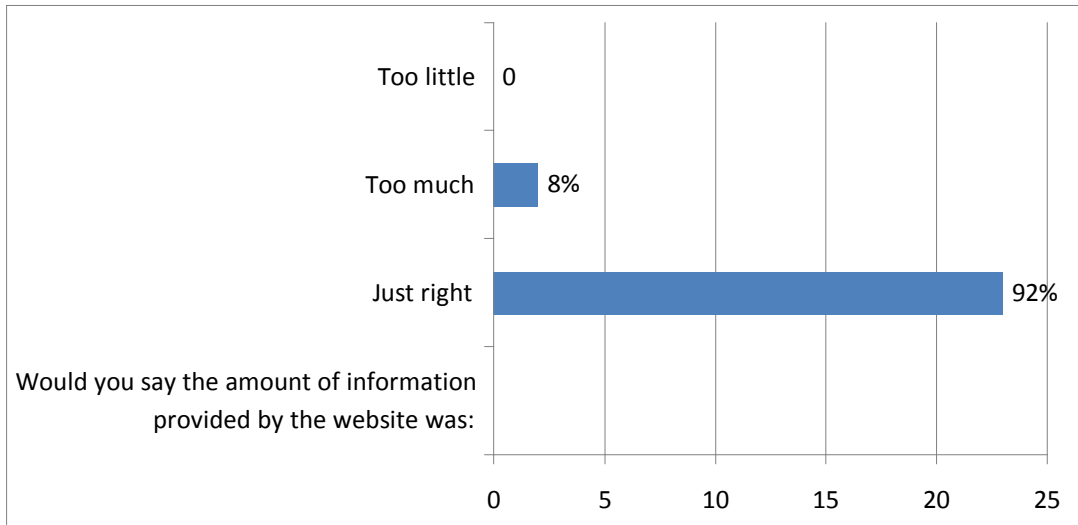


Figure 8. Satisfaction with amount of information on website

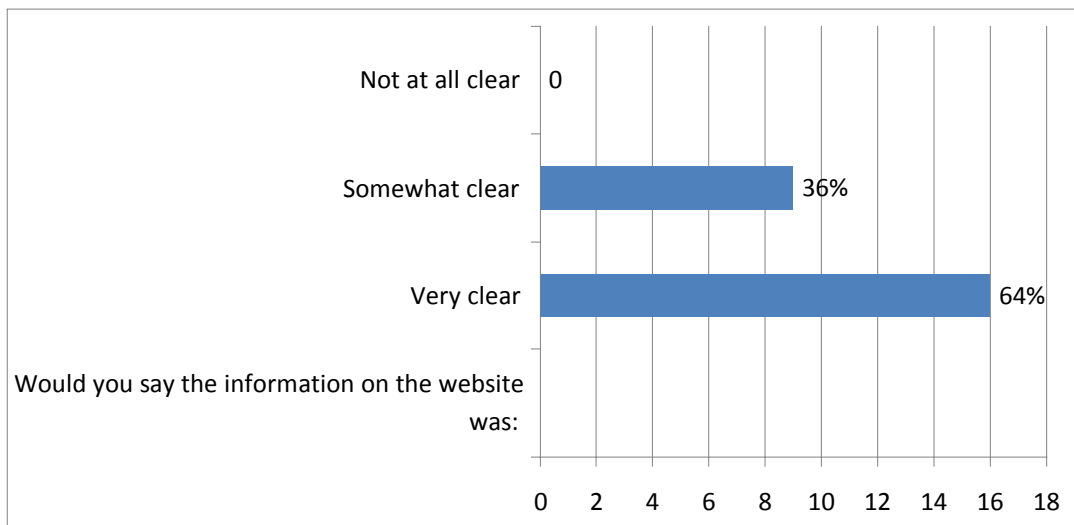


Figure 9. Clarity of Information on Website

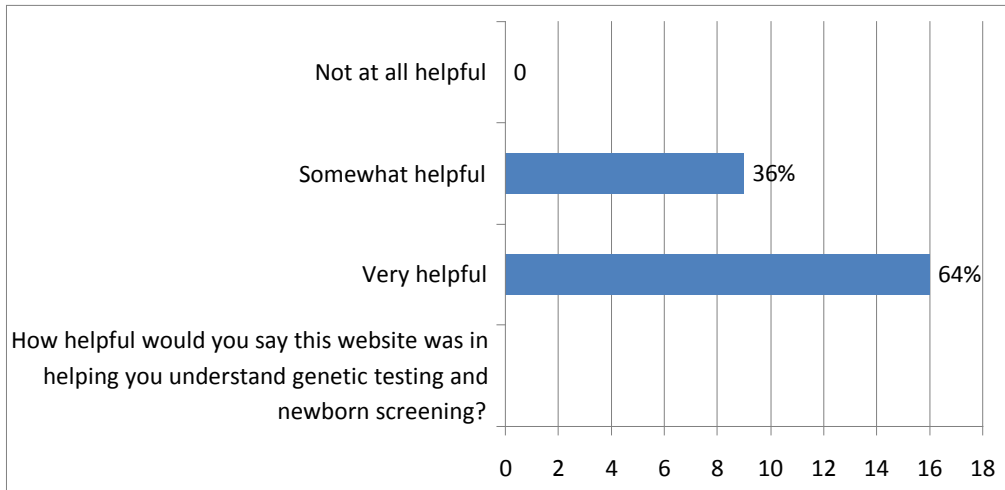


Figure 10. Helpfulness of Website in Understanding Genetic Testing and NBS

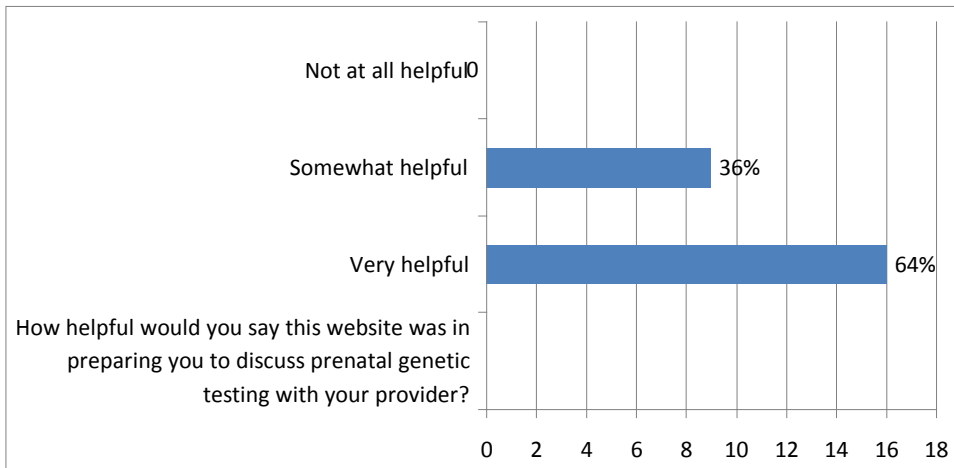


Figure 11. Helpfulness in Discussing Genetic Testing with Provider

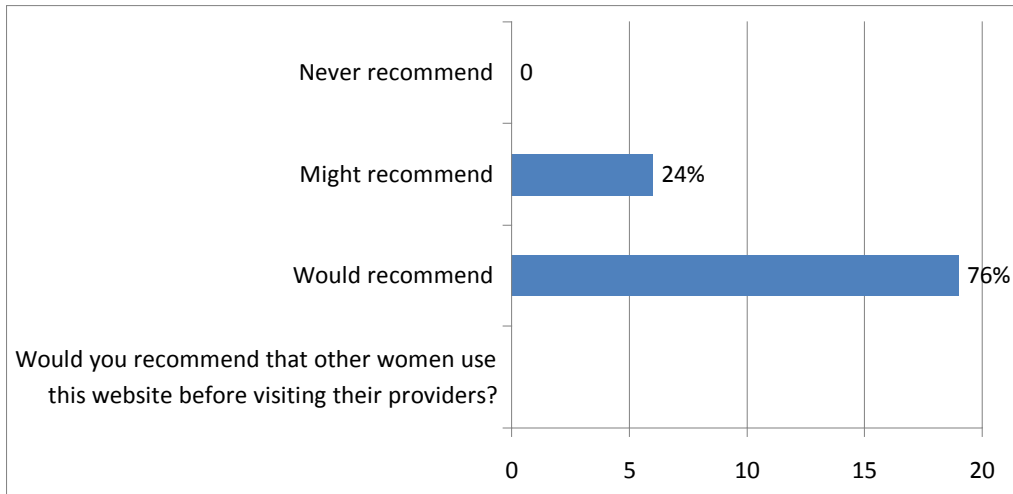


Figure 12. Recommending Website to Others

5.0 DISCUSSION

5.1 SPECIFIC AIM 1

The aim of implementing this website into the workflow of the clinic had limited success. The clinic has a high volume of patients, which made it challenging to incorporate this website consistently into patients' clinic visits. For this study, patients were shown the website at the end of their time in clinic, after they had seen all their providers and had their blood drawn in the lab. This is not the ideal time for them to see the website, since they would have already seen their prenatal care providers and were frequently tired from spending a few hours in clinic. It would have been preferred for a patient to read through the website before meeting with her obstetrician, since she would then have the opportunity to ask questions and it could potentially cut down on the provider's counseling time.

5.2 SPECIFIC AIM 2

The effectiveness of this website was successfully evaluated. The data indicate that it is helpful in improving women's knowledge of genetics and newborn screening, shown both by improvement in knowledge-based questions from a pre- to post-test and by opinion-based questions. On average, participants answered as many questions correctly on the post-test, and

their feedback was overwhelmingly positive. All participants believe the website was at least somewhat helpful in helping them understand genetic testing and newborn screening. Differences in race, educational level, and relationship status had no statistically significant impact on a participant's score. The fact that there was no evidence of a culture bias in the website is important, particularly for the African-American population since there is a history of mistrust of medical professionals [25]. It has been shown that younger individuals have a general wariness of physicians, even though they have never heard of events such as the Tuskegee Syphilis study. It has been suggested that this sentiment has been ingrained into the African American culture and is passed down through generations [25]. When there is mistrust of health care providers, a website reinforcing information could be useful as an additional means of education.

The only factors that had a significant effect on score were the participant's age and number of previous children. Age had an inverse correlation with score on the post-test, but had no impact on performance on the pre-test. All participants were relatively young (mean age of 22 years), therefore it seems unlikely that older individuals would have less experience with technology so that their scores would be lower. This analysis was re-calculated to adjust for previous children, and the age difference was still present. A larger sample size with a balance of older participants could be helpful to determine how significant this difference is.

The number of previous children had an impact on pre-test score, but not on the post-test. The pre-test score was higher for those with other children, but they did not score any better on the post-test. This suggests that mothers had some familiarity with newborn screening and carrier testing from previous pregnancies. Since there was no difference in post-test scores, it

could be that reading the website brings the new moms' knowledge up to the level of someone with previous children.

5.3 STUDY LIMITATIONS

Though the findings of this study have allowed this educational tool to be evaluated, there are some limitations. One of the main limitations is that those who were Caucasian did not see information on sickle cell disease while reading the website, but two of the survey questions were on sickle cell disease. Therefore, these individuals were being tested on information to which they weren't exposed. The sample size of 25 participants was relatively small, and more participants could have provided additional strength to the findings. It was also difficult to assess if this website is a practical tool for this clinic, since patients never went to the website unless they were approached and personally led to it. It was a challenge to implement this website into the natural flow of the clinic and for patients to view the information at the most opportune time.

5.4 AREAS FOR FUTURE STUDY

Given that there was difficulty in implementing the website in the clinic, further study could be done to evaluate the website in a different clinical setting, such as a private practice. If a clinic has success in incorporating the website so that women see it prior to meeting with their

physician, it would be interesting to compare how much having read the website affects their conversation with the obstetrician. Once the website has become consistently utilized in a clinic, individuals who either test positive on a carrier screen or have a newborn with a positive screening result can be surveyed to determine if they were familiar with My Baby's Health, and if learning about NBS and carrier testing in the prenatal period was helpful. Additional analysis could be done to determine if people who found the website "somewhat helpful" or "somewhat clear" did more poorly on the post-test. Separate surveys could also be developed for those who are Caucasian so that this population is not tested on information they did not see while reading the website.

5.5 CONCLUSION

The data indicate that the My Baby's Health website is a useful tool in improving knowledge on carrier testing and newborn screening. This website would be most beneficial if it were incorporated into the natural flow of the clinic, so that all prenatal patients are encouraged to visit the website while waiting to meet with the provider. Exposure to carrier testing and newborn screening beforehand could reduce the time a provider would need to spend counseling on that information. Since increased patient knowledge of carrier testing and NBS has been suggested to improve follow-up, this website can help newborns who have tested positive to receive more timely management. Improved education also has the benefit of reducing anxiety that accompanies a positive newborn or carrier screening test. If utilized to its full potential in numerous prenatal clinics across the United States, the My Baby's Health website could have public health impact. It could decrease health disparities across the country by making education

on newborn screening and carrier testing more accessible to a wider array of individuals, including those of different socioeconomic and ethnic backgrounds.

APPENDIX A: SURVEYS

A.1 PRE-WEBSITE SURVEY

Dear Viewer,

The purpose of this research study is to determine whether a website can effectively provide education about genetics and testing to pregnant women. We plan to survey women who have an appointment with a prenatal care provider during their pregnancy. The two questionnaires are brief (totaling 5-10 minutes) and will be presented before and after the website is viewed.

If you are willing to participate, our questionnaire will ask you about genetic testing and genetic diseases, as well as about your background (e.g., age, education, race). There are no foreseeable risks associated with this survey. There are no direct benefits to you and you will not receive payment for participating. Your participation is voluntary, and you may withdraw at any time.

This study is being conducted by Dr. Lakshmanan Krishnamurti, a pediatric hematologist, and Claire Harwood, BA, a genetic counseling intern. The study personnel can be reached at 412-692-7827, if you have any questions.

Genetics and Testing

These 7 questions are about what you may have heard about genetic conditions and testing.

1. Have you heard of any of the following conditions or tests? Check all that apply.

- ☐ Sickle cell trait
- ☐ Sickle cell disease
- ☐ Cystic fibrosis (CF)
- ☐ Thalassemia
- ☐ Newborn screening

☐ I have not heard of any of these

2. A positive sickle cell carrier test means:

- ☐ That person definitely has sickle cell trait
- ☐ That person probably has sickle cell trait
- ☐ That person could develop sickle cell trait over time
- ☐ There is no test for sickle cell trait
- ☐ Don't know

3. How can a child get sickle cell disease?

- ☐ Both parents must have sickle cell trait
- ☐ Their mom has sickle cell trait but their dad does not
- ☐ Their dad has sickle cell trait but their mom does not
- ☐ One parent also has sickle cell disease
- ☐ Don't know

4. A negative cystic fibrosis carrier test means:

- ☐ That person is definitely not a carrier
- ☐ That person is probably not a carrier, although this cannot be definitely ruled out
- ☐ There is no carrier test for cystic fibrosis
- ☐ Don't know

5. If two parents are carriers for cystic fibrosis, they can have healthy children.

- ☐ True
- ☐ False
- ☐ Don't know

6. How is newborn screening performed?

- ☐ The baby goes to a check-up with a doctor
- ☐ The baby's blood is drawn by pricking their heel
- ☐ The baby's blood is drawn from their arm
- ☐ The baby's DNA is taken from their saliva
- ☐ Don't know

7. A healthy baby can receive a positive (abnormal) newborn screening result.

- ☒ True
- ☐ False
- ☐ Don't know

A.2 POST-WEBSITE SURVEY

These 6 questions are about what you may have heard about genetic conditions and testing.

A positive sickle cell carrier test means:

- ☒ That person definitely has sickle cell trait
- ☐ That person probably has sickle cell trait
- ☐ That person could develop sickle cell trait over time
- ☐ There is no test for sickle cell trait
- ☐ Don't know

How can a child get sickle cell disease?

- ☒ Both parents must have sickle cell trait
- ☐ Their mom has sickle cell trait but their dad does not
- ☐ Their dad has sickle cell trait but their mom does not
- ☐ One parent also has sickle cell disease
- ☐ Don't know

A negative cystic fibrosis carrier test means:

- That person is definitely not a carrier
- That person is probably not a carrier, although this cannot be definitely ruled out
- There is no carrier test for cystic fibrosis
- Don't know

If two parents are carriers for cystic fibrosis, they can have healthy children.

- True
- False
- Don't know

How is newborn screening performed?

- The baby goes to a check-up with a doctor
- The baby's blood is drawn by pricking their heel
- The baby's blood is drawn from their arm
- The baby's DNA is taken from their saliva
- Don't know

A healthy baby can receive a positive (abnormal) newborn screening result.

- True
- False
- Don't know

These 5 questions tell us how you felt about the MyBabysHealth.org website.

Would you say the amount of information provided by the website was: *

- Too little
- Just right
- Too much

Would you say the information on the website was:

- Not at all clear
- Somewhat clear
- Very clear

How helpful would you say this website was in helping you understand genetic testing and newborn screening?

- Not at all helpful
- Somewhat helpful
- Very helpful

How helpful would you say this website was in preparing you to discuss prenatal genetic testing with your provider?

- Not at all helpful
- Somewhat helpful
- Very helpful

Would you recommend that other women use this website before visiting their providers?

- Never recommend
- Might recommend
- Would recommend

APPENDIX B: IRB-APPROVAL



University of Pittsburgh
Institutional Review Board

3500 Fifth Avenue
Pittsburgh, PA 15213
(412) 383-1480
(412) 383-1508 (fax)
<http://www.irb.pitt.edu>

Memorandum

To: Dr. Lakshmanan Krishnamurti
From: Sue Beers, PhD, Vice Chair
Date: 2/14/2012
IRB#: PRO12020002
Subject: Evaluation of the Impact of a Web-Based Educational Tool on Awareness of Newborn Screening

The above-referenced project has been reviewed by the Institutional Review Board. Based on the information provided, this project meets all the necessary criteria for an exemption, and is hereby designated as "exempt" under section 45 CFR 46.101(b)(2).

The IRB has approved the advertisement that was submitted for review as written. As a reminder, any changes to the approved advertisement would require IRB approval prior to distribution.

Please note the following information:

- If any modifications are made to this project, use the "**Send Comments to IRB Staff**" process from the project workspace to request a review to ensure it continues to meet the exempt category.
- Upon completion of your project, be sure to finalize the project by submitting a "**Study Completed**" report from the project workspace.

Please be advised that your research study may be audited periodically by the University of Pittsburgh Research Conduct and Compliance Office.

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